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Interventional Cardiology

Management of Coronary Artery Fistulae

Patient Selection and Results of Transcatheter Closure

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OBJECTIVES We report short-term findings in 33 patients after transcatheter closure (TCC) of coronary artery fistulae (CAF) and compare our results with those reported in the recent transcatheter and surgical literature.

BACKGROUND Transcatheter closure of CAF has been advocated as a minimally invasive alternative to surgery.

METHODS We reviewed all patients presenting with significant CAF between January 1988 and August 2000. Those with additional complex cardiac disease requiring surgical management were excluded.

RESULTS Of 39 patients considered for TCC, occlusion devices were placed in 33 patients (85%) at 35 procedures and included coils in 28, umbrella devices in 6 and a Grifka vascular occlusion device in 1. Post-deployment angiograms demonstrated complete occlusion in 19, trace in 11, or small residual flow in 5. Follow-up echocardiograms (median, 2.8 years) in 27 patients showed no flow in 22 or small residual flow in 5. Of the 6 patients without follow-up imaging, immediate post-deployment angiograms showed complete occlusion in 5 or small residual flow in 1. Thus, complete occlusion was accomplished in 27 patients (82%). Early complications included transient ST-T wave changes in 5, transient arrhythmias in 4 and single instances of distal coronary artery spasm, fistula dissection and unretrieved coil embolization. There were no deaths or long-term morbidity. Device placement was not attempted in 6 patients (15%), because of multiple fistula drainage sites in 4, extreme vessel tortuosity in 1 and an intracardiac hemangioma in 1.

CONCLUSIONS A comparison of our results with those in the recent transcatheter and surgical literature shows similar early effectiveness, morbidity and mortality. From data available, TCC of CAF is an acceptable alternative to surgery in most patients. (J Am Coll Cardiol 2002;39: 1026–32) © 2002 by the American College of Cardiology Foundation

Coronary artery fistulae (CAF) are rare anomalies, commonly diagnosed through evaluation of a continuous murmur in an asymptomatic child. Elective closure in childhood has been recommended (1–3) after reports that patients with CAF may develop complications such as myocardial ischemia, congestive heart failure, endocarditis or aneurysmal dilation during or after the second decade of life (4,5).

Transcatheter closure (TCC) of CAF has been utilized since 1983 (4,6–15). Prior reports, including our own (14), have been limited in patient number (2 to 13 patients) and follow-up data (longest mean follow-up, 1.5 years). Beginning in 1988, our institutional approach has been to attempt TCC in all patients presenting with clinically significant, isolated CAF. We present our experience to: 1) describe the techniques used in TCC of CAF; 2) report our results with this procedure; 3) compare our findings with those described in the transcatheter and recent surgical literature; and 4) outline our current indications for surgical or TCC of CAF.

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METHODS

Patient identification, selection and classification. From our medical and surgical databases, we identified all patients with CAF who underwent a catheterization or surgical procedure between January 1988 and August 2000. After exclusion of those with additional complex cardiac disease requiring surgical management, and of those with tiny CAF found incidentally during echocardiography or catheterization, we identified 39 patients with clinically significant CAF (i.e., those producing symptoms or a typical murmur). Of the 39 patients, TCC was undertaken in 33 (TCC patient group) but not attempted in 6 patients (non-intervention patient group).

Clinical, electrocardiogram and echocardiographic assessment. All patients underwent a cardiac evaluation, including medical history, physical examination, electrocardiogram (ECG) and echocardiogram. Echocardiographic features indicating a significant CAF included (one or more) the following: cardiac chamber dilation; narrowest color Doppler flow jet ≥4 mm; or reversal of flow in the descending aorta.

Abbreviations and Acronyms

CAF = coronary artery fistulae
Cx = circumflex
ECG = electrocardiogram
LAD = left anterior descending
LCA = left coronary artery
LV = left ventricle
RA = right atrium
RCA = right coronary artery
RV = right ventricle
TCC = transcatheter closure

Transcatheter closure technique. Femoral arterial and venous catheters were placed via percutaneous access. After collection and analysis of hemodynamic data, the anatomy and dimensions of the coronary arteries and fistulae were determined using selective (often power-assisted) coronary angiograms. Hand injections of contrast facilitated catheter positioning in the distal fistula. Due to the typically high flow rates in CAF, balloon occlusion angiography was used within the fistula to precisely identify all distal coronary branches, fistula dimensions and drainage sites. Catheter positioning within the distal fistula was often facilitated by the use of high-torque floppy wires or tracker catheters (Tracker Infusion Catheter, Medi-tech/Boston Scientific Corp., Watertown, Massachusetts), as reported by other investigators (16,17).

Feasibility of closure by a device was determined by the number and location of drainage sites, the ability to cannulate the distal fistula and the proximity of coronary branches to the optimal occlusion site. Special care was taken to avoid allowing the device to interfere with flow into any visible coronary artery. Occlusion devices included Gianturco coils (Occluding Spring Emboli; Cook Cardiology; Bloomington, Indiana), double umbrella devices (Bard PDA Umbrella, Bard Clamshell Septal Umbrella, USCI Division, C.R. Bard; CardioSEAL: Nitinol Med Techn Inc.; Boston, Massachusetts) and Gianturco Grifka vascular occlusion device (GGVOD: Cook Cardiology; Bloomington, Indiana). The selection of occlusion device was based on the anatomic features of the fistula. Coils, used primarily in smaller CAF (Fig. 1), offer advantages of smaller sheath and catheter delivery sizes and lower cost. Double umbrella devices allow more precise positioning and were used in larger fistulae with coronary branches close to the occlusion site.

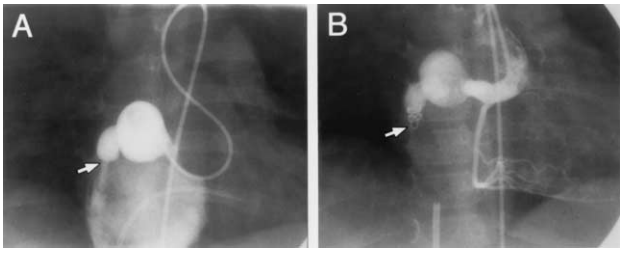

Figure 1. Coronary artery fistula from right coronary artery to right atrium. **(A)** Single insertion site into right atrium **(arrow)**. **(B)** Complete occlusion following transvenous placement of single coil **(arrow)**.

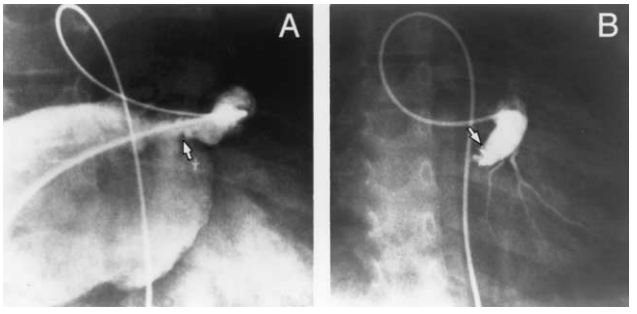

Figure 2. Coronary artery fistula from left coronary artery to left atrium. **(A)** Arteriovenous wire loop enabling passage of venous catheter across the atrial septum into the fistula drainage site **(arrow)**. **(B)** Angiogram following transvenous deployment of a 12-mm Rashkind device **(arrow)**, showing coronary artery fistula occlusion and coronary artery side branches that were not evident in angiograms performed without balloon occlusion.

To occlude the distal fistula immediately proximal to the drainage site, we selected coils 20% to 40% larger than the maximum diameter of the vessel. To occlude the drainage orifice into a cardiac chamber by straddling this site, we selected coils or double umbrella devices approximately twice the diameter of that orifice. Device deployment was performed either antegrade (via the femoral vein) or retrograde (via the femoral artery). Antegrade deployment avoids potential damage to the femoral artery, allows use of larger catheters and affords a straighter catheter course. An arteriovenous wire loop to allow TCC from either approach (Fig. 2) was created where feasible by first guiding a catheter through the fistula via the aorta, then passing a wire via this catheter into the right heart, where it was snared and extracted via the femoral vein.

Follow-up. Medical records including echocardiograms and ECGs were reviewed. The presence and degree of residual flow after TCC were determined by the most recent echocardiographic data. When follow-up echocardiograms were not performed, the most recent clinical data in conjunction with the angiographic results at the conclusion of TCC were used.

Data analysis. Medical records including clinical evaluations, catheterization, ECG and echocardiographic and follow-up data were reviewed in all patients. Approval to conduct the medical record review was obtained from our Institutional Review Board.

RESULTS

Transcatheter closure patient group. Thirty-nine patients (age, 8 months to 68 years; median age, 8 years) underwent cardiac catheterization with the intent to close the CAF. Occlusion devices were placed in 33 patients (85%). Of these, two patients underwent a second catheterization for significant residual fistulae, resulting in a total of 35 procedures. Additional cardiac lesions were present in six patients, consisting of atrial arrhythmias in two patients and single instances of pulmonary atresia post Fontan procedure,

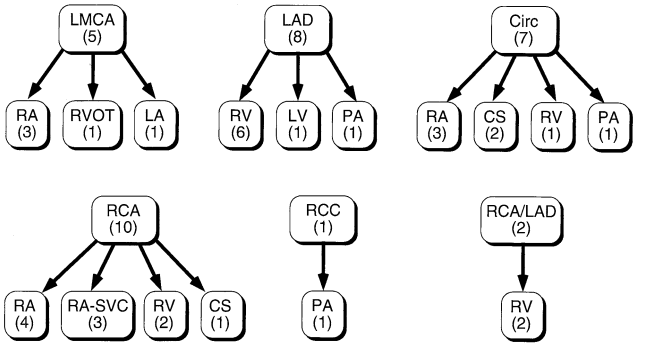

Figure 3. The transcatheter closure patient group: coronary artery fistulae origin and drainage sites. Circ = circumflex artery; CS = coronary sinus; LA = left atrium; LAD = left anterior descending coronary artery; LMCA = left main coronary artery; LV = left ventricle; PA = pulmonary artery; RA = right atrium; RCA = right coronary artery; RCC = right coronary cusp; RV = right ventricle; RVOT = right ventricular outflow tract; SVC = superior vena cava.

aortic stenosis post Ross procedure, post heart transplant and a single left coronary artery (LCA). Thirteen patients (39%) were symptomatic (dyspnea on exertion, 8; chest pain, 3; palpitations, 1; and fatigue, 1). Continuous murmurs were present in 31 patients and systolic murmurs in 2. Pre-catheterization ECGs were normal in 25 of the 33 patients (76%), with left ventricular (LV) hypertrophy evident in 4, non-specific ST-T wave changes in 3 and right atrial (RA) enlargement in 1.

The sites of CAF origin and drainage are detailed in Figure 3. Origin from the LCA was more common than from the right, and drainage to the right heart occurred in all but two. Fistulae originated from two coronary arteries in two patients and had multiple drainage sites in three others.

At 35 catheterizations (33 patients), coils were placed in 28 procedures, Rashkind or CardioSEAL devices in 6, and a Grifka vascular occlusion device in the other (Fig. 4). Nineteen devices were deployed retrograde and 16 antegrade; an arteriovenous wire loop facilitated antegrade delivery in 11 patients. In five patients, tracker catheters facilitated distal positioning of coils within long and tortuous fistulae.

Immediate post-deployment angiograms (Fig. 5) demonstrated complete occlusion in 19, trace (barely perceptible) flow in 11 and small residual flow in five. A second catheterization for TCC of significant residual fistulae was performed in two patients; in the first, angiography showed trace residual flow immediately after placement of a 12-mm umbrella device; but six months later, a continuous murmur was noted, and echocardiography revealed significant residual flow. At re-catheterization, eight coils placed within the fistula resulted in complete occlusion both angiographically and on follow-up echocardiography. In the second patient, implantation of two 17-mm CardioSEAL devices at two drainage sites into the RA resulted in trace residual flow angiographically. A continuous murmur was noted at follow-up, and echocardiography showed significant residual flow. At re-catheterization, 11 coils placed within the fistula resulted in trace residual flow by angiography and follow-up echocardiography. In both patients the continuous murmurs were absent at follow-up.

Procedural complications included transient ST-T wave changes in five, transient atrial arrhythmia in four, and single instances of distal coronary artery spasm with contrast injection, unretrieved embolized coil to a pulmonary artery and fistula dissection. In the latter, the dissection occurred at the site of coil implantation, was followed by fistula thrombosis and was without sequelae. There were no deaths, strokes or infections. Electrocardiograms were available after 29 procedures and were unchanged from baseline

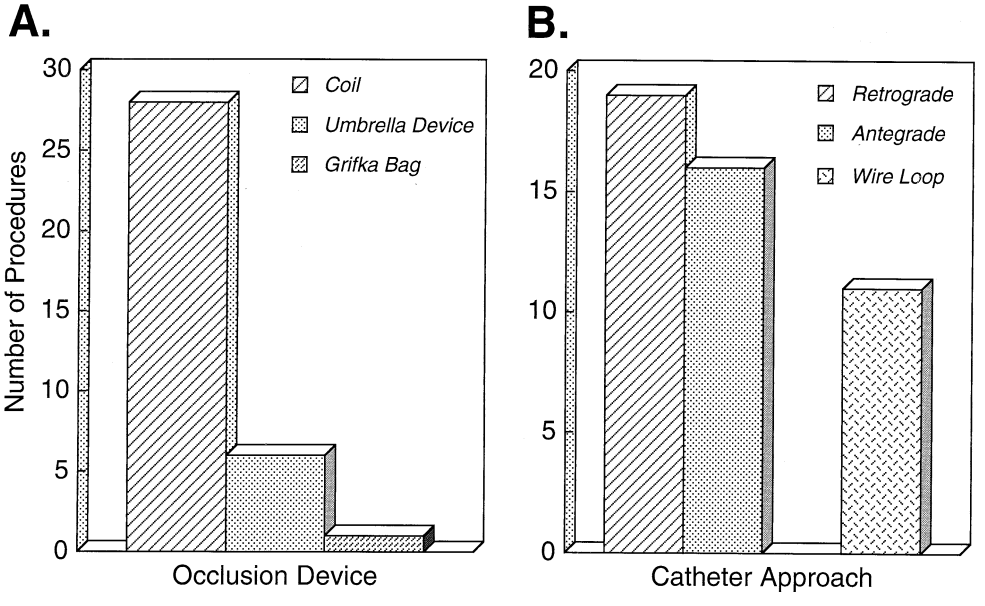

Figure 4. Device delivery in the transcatheter closure patient group. **(A)** Occlusion device used. **(B)** Catheter approach for device delivery.

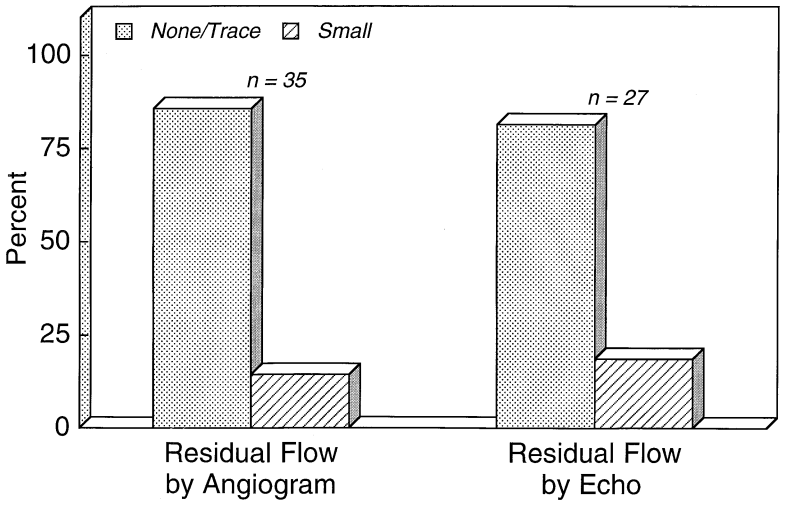

Figure 5. Residual flow following transcatheter closure of coronary artery fistula by angiography and echocardiography.

in 24 with transient ST-T wave changes noted in five patients.

Non-intervention patient group. Placement of an occlusion device was not attempted in 6 (15%) of the 39 patients initially considered for TCC, all but one catheterized early in our experience (1988 to 1993). These patients ranged in age from 3 weeks to 48 years (median age, 4 years) at catheterization; additional cardiac lesions were present in three patients, consisting of a single LCA in two and intracardiac hemangioma in the other. One patient was symptomatic; all had continuous murmurs. The sites of CAF origin and drainage are detailed in Table 1. Placement of a device was not attempted due to the presence of multiple fistula drainage sites in four, extreme vessel tortuosity with inability to cannulate the distal fistula (Fig. 6) in one and the presence of multiple minute drainage sites from an intracardiac hemangioma in the other. There were no immediate or late complications.

Follow-up. TCC PATIENT GROUP. At follow-up (median, 2.8 years; range, 1 month to 11.1 years), all patients were asymptomatic, and 30 were without continuous murmurs. The most recent echocardiograms, available in 27 patients, identified complete occlusion in 22 (82%) and small residual flow in 5 (18%) (Fig. 5). Six patients have not had follow-up echocardiograms; of these, five had no residual flow by angiography immediately after device placement, and the other had a small residual shunt and a persistent continuous murmur. Thus, complete occlusion was accomplished in 27 patients (82%).

NON-INTERVENTION PATIENT GROUP. Follow-up data (median, 7.4 years; range, 5.8 to 11 years) were available in only three of the six patients. They were asymptomatic: the murmur had disappeared in the patient with an intracardiac hemangioma, and echocardiograms, available in only two patients, were unchanged.

DISCUSSION

Elective closure of significant CAF in childhood has been advocated to prevent later complications (1,5,8,11). Since its introduction in 1983 (6), TCC of these lesions is increasingly being utilized as an alternative to surgical closure.

Transcatheter and surgical literature review. TCC LITERATURE. Excluding single case reports, TCC of CAF has been reported in 45 patients (5 weeks to 71 years of age; median, 12 years) since 1982 (4,7–13,15) (Table 2). The data from our previously published report (14) are included in our current patient group. The fistulae were isolated lesions in all but one (prior aortic valve replacement). Symptoms were reported in 18 (40%), including angina in 10, dyspnea on exertion in 6 and single instances of fatigue and poor growth. The fistulae originated from the LCA in

Table 1. Transcatheter Non-Intervention Patient Group

Patient	Age	Fistula Origin	Fistula Drainage	Reason for No Intervention
1	48.6 yrs	RCA	CS	Vessel tortuosity
2	4.2 yrs	LAD	RV	Multiple drainage sites
3	1.2 yrs	LAD	RV	Multiple drainage sites
4	20 days	RCA/LCA	RA	Hemangioma
5	5.0 yrs	RCA	RA	Multiple drainage sites
6	4.0 yrs	LAD/Cx	RV	Multiple drainage sites

CS = coronary sinus; Cx = circumflex artery; LAD = left anterior descending artery; LCA = left coronary artery; RA = right atrium; RCA = right coronary artery; RV = right ventricle.

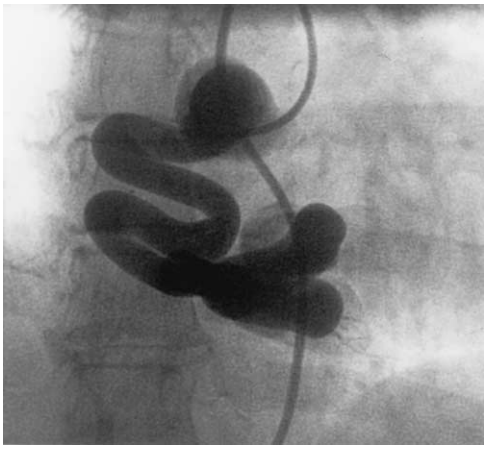

Figure 6. Angiogram showing tortuous coronary artery fistula from right coronary artery to coronary sinus (Table 1, Patient 1): occlusion not attempted as antegrade access to coronary artery fistula unsuccessful and catheter lengths insufficient to reach distal fistula from a retrograde approach.

24 (10 from the left anterior descending [LAD], seven from the circumflex [Cx]), from the right coronary artery (RCA) in 19, and from both RCA/LCA in two. Fistulae drained to the RA in 14, right ventricle (RV) in 12, pulmonary artery in 11, coronary sinus in 3, LV in 2, bronchial artery in 2 and superior vena cava in 1.

Transarterial device delivery was performed in 41 procedures. Coils were placed in 31, detachable balloons in 10, umbrella devices in 2, covered stent in 1 and a combination of detachable balloons and coils in 2. Successful occlusion of the CAF at catheterization was reported in 83% of patients. One patient required re-catheterization for complete closure. Procedural complications included transient ischemic changes on ECG in four, unretrieved device embolization to tricuspid valve or distal pulmonary artery in a similar number and single instances of myocardial infarction and transient atrial arrhythmia. There was one procedure-related death (2.2%) involving a symptomatic 71-year-old man with a large CAF arising from a diffusely diseased LAD and inserting into a pulmonary artery. Death resulted from embolization of a coil to the LCA and subsequent LCA dissection (4).

At follow-up (1 day to 4 years; longest mean follow-up, 12 months) in 42 of 45 patients, all were asymptomatic and without late complications or deaths. At follow-up imaging studies of 33 patients (echocardiograms in 27, angiograms in 6), complete closure was reported in 91%.

SURGICAL LITERATURE. Surgical series published in the past decade (1–3,18–20) consisted of 71 patients (81 fistulae) (Table 2). These patients ranged in age from 6 days to 77 years (median, 6 years); 19 had additional cardiac lesions (27%), including patent ductus arteriosus in 4, atrial septal defect and mitral regurgitation in 3 each, 2 patients each with aortic regurgitation, tetralogy of Fallot, ventricular septal defect, RCA origin atresia and coarctation of the aorta in 1. Closure of CAF was performed concurrent with additional cardiac surgical procedures in 11 patients (15%), including valve replacement, atrial or ventricular septal defect repair and patent ductus arteriosus ligation. The use of cardiopulmonary bypass was reported in 37 patients (57%). The fistulae arose from the RCA in 21 patients, the LCA in 40 (LAD in 18, Cx in 7) and both RCA/LCA in 10. Drainage sites included the RV in 28 patients, pulmonary artery in 20, RA in 18, RV outflow tract in 3 and 1 each in the left atrium and LV. There was one operative death (1.4%) secondary to myocardial infarction (19). Other complications included arrhythmia in five, transient ischemic changes on ECG in one and stroke in another. There were no reported late post-operative deaths. Follow-up was available in 65 patients (range, 0.1 to 21.1 years; longest mean follow-up, 7.2 years), and all were asymptomatic. Follow-up imaging studies were reported in only 21 (30%) of the 70 survivors (angiography, 13; echocardiography, 8), with complete closure rates in these ranging from 50% to 100% (1,18,20).

Observations. The origin and drainage sites in the TCC and surgical literature are similar to our data except for our lower incidence of drainage to a pulmonary artery. Delivery of the occlusion device (usually coils) was more often performed via a retrograde approach in the transcatheter literature (89%) compared with our series (54%). Follow-up imaging was available in 75% of the patients reported in the transcatheter literature, 30% of surgical patients and 82% of patients in our series. Because of this wide disparity in follow-up imaging data, a valid comparison of documented closure of CAF between the techniques is difficult. However, given the available data, complete closure by transcatheter and surgical techniques are comparable, as are rates of early and late complications (Table 2).

While we have found TCC of CAF feasible in most patients, features that may render CAF unsuitable for this technique include those with extreme vessel tortuosity, multiple drainage sites and coronary branches at the site of optimal device positioning. The ability to cannulate the distal fistula and avoid flow interference through nearby coronary branches is mandatory for successful TCC. Early in our experience (prior to 1994), we determined that CAF with multiple drainage sites precluded an attempt at TCC; however, with experience and improvements in catheterization equipment and occlusion devices, we have subsequently successfully closed fistulae with this characteristic in three patients.

An "ideal" study to provide a rigorous comparison of transcatheter versus surgical therapy would necessitate a randomized multicenter trial with fixed entry criteria, similar operator skills at each of the institutions and an extended period of follow-up. The technique of therapy would need to remain largely fixed during the course of the study. Without the data from such a trial, we have adopted the following therapeutic strategy: 1) those with CAF and additional complex heart disease requiring surgery are referred for surgical repair; 2) patients with a clinically

Table 2. Current Data and Literature Review

	Study Dates	Patients	Median Age (yrs)	Isolated Fistulae	Morbidity	Mortality	Mean Follow-up (yrs)	Residual Fistulae
Surgical literature	1968–1996	71	6	52 (73%)	Arrhythmia (5) Stroke (1) Transient ischemic changes (1) (10%)	1 (1.4%)	7.2* (0.1–21.1 yrs)	6 (24 f/u studies) (25%)
TCC literature	1982–2000	45	12	44 (98%)	Unretrieved coil embolization (4) Transient ischemic changes (4) Infarction (1) Transient arrhythmia (1) (22%)	1 (2.2%)	1.0* (1 d–4 yrs)	3 (33 f/u studies) (9%)
Current TCC report	1988–2000	33	8	26 (79%)	Transient ST-T changes (5) Transient arrhythmia (4) Distal coronary artery spasm (1) Fistula dissection (1) Unretrieved coil embolization (1) (34%)	0	3.7 (0.1–11.1 yrs)	5 (27 f/u studies) (19%)

*Longest mean follow-up.
f/u = follow-up; TCC = transcatheter closure.

significant CAF (i.e., producing symptoms or a typical murmur) are referred for catheterization, where the fistula anatomy is further defined; 3) those with suitable anatomy undergo TCC, while those unsuitable for TCC are offered surgical ligation or are followed up medically.

With regard to tiny CAF now being diagnosed with improved color flow Doppler technology, we currently, as do other investigators (20,21), follow such patients without intervening, because the incidence of cardiac complications is low and there is the possibility of spontaneous closure. There are others, however, who disagree with this strategy (5,22).

Study limitations. A meaningful comparison of transcatheter and surgical closure of CAF is limited by (a) small patient numbers and (b) the paucity of follow-up imaging, especially in the surgical literature. Additionally, although we have limited our review of the surgical literature to series that were published within the past decade, each of these includes patients treated prior to this period.

CONCLUSIONS

With increased experience and improved devices and techniques, TCC of CAF is emerging as a successful therapeutic strategy. The safe and effective results of both surgical closure and TCC support the current convention of elective closure of clinically significant CAF in childhood. The preferred method of approach for any individual patient will depend on the anatomy of the fistula, the presence or absence of associated defects and the experience of the interventional cardiologists and surgeons.

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